

# Hemangiolympangioma of buccal mucosa: A rare case report

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## Abstract

Vascular anomalies are mainly classified under two headings, i.e. under vascular tumors and vascular malformations. Hemangioma and lymphangioma are examples for such a vascular anomalies. Malformations may be seen in different combinations of vascular elements, and histologically these vessels may be filled with blood and named lymphangiohemangioma or hemangiolympangioma according to the dominant tissue structure present. It is a rare developmental anomaly. This paper reports a case of lymphangiohemangioma in a 21-year-old female patient.

**Keywords:** Hemangioma, lymphangiohemangioma, lymphangioma, vascular anomalies

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## INTRODUCTION

Vascular anomalies are mainly classified under two headings, i.e. under vascular tumors and vascular malformations according to the cellular turnover, histology and clinical findings.<sup>[1]</sup> Hemangioma is usually seen during infancy and childhood, occurring in 4%–10% of Caucasian infants. Hemangiomas are generally noted within the first 2 weeks of postnatal life. However, they are wide variability in this timing.<sup>[2]</sup> Hemangiomas mostly occur in the female. The majority of hemangiomas involve the head and neck. However, there are rare in the oral cavity.<sup>[3]</sup> The hemangioma may present with a macular, telangiectatic appearance. They are classified mainly cavernous and capillary types.<sup>[4]</sup> It could be seen in the cutaneous, mucosal, intramuscular or intraosseous.<sup>[5,6]</sup>

Lymphangioma is considered as a benign hamartomatous tumor of lymphatic vessels. It is widely considered as

a developmental and congenital lesion rather than a true neoplasia.<sup>[1]</sup> Lymphangiomas are usually noted at birth or within 2 years of life. Most commonly occur in the cervicofacial region, axilla/chest, mediastinum, retroperitoneum, buttock and perineum. The overlying skin is usually normal or may have a bluish hue.<sup>[2]</sup> Lymphangioma is classified by the diameter of the vessels into capillary lymphangioma, cavernous lymphangioma and cystic lymphangioma.<sup>[1]</sup>

Malformations may be seen in different combinations of vascular elements, such as lymphatic and venous endothelium and cannot be identified as purely one or the other entities. Histologically these vessels may be filled with blood, a mixed hemangiolympangioma and a rare developmental anomaly. These mixed vascular malformations are named as lymphangio-hemangioma or hemangiolympangioma according to the dominant

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tissue structure.<sup>[6-9]</sup> Here, we present a patient with a hemangiolympangioma of the buccal mucosa.

## CASE REPORT

A 21-year-old female patient reported to the Department of Oral Medicine and Radiology of the Mahe Institute of Dental Sciences and Hospital, Mahe, Union Territory of Puducherry, India, in the month of March 2016, with the complaint of reddish bluish swelling over the left inner side of cheek after an accidental cheek bite 7 days back. The patient gave a history of similar episodes of cheek biting and resultant bleeding occasionally. No history of gross change in the size of the swelling was reported. Patient's mother had consulted a pediatric surgeon when she was 5 years of age after noticing mild swelling of the left side of her face. She was advised to be kept on follow-up and suggested that the lesion would regress with age. Past medical and family history was noncontributory. Extraoral examination revealed slight facial asymmetry on the left side [Figure 1] with asymmetric nasolabial folds on smiling. Facial muscle movements were normal; however, the imbalance in facial animation was apparent. On intraoral inspection, a solitary reddish bluish swelling with the translucent yellow surface was seen on the left buccal mucosa measuring approximately 2 cm × 1 cm in dimension extending superior-inferiorly from the occlusal plane, 1 cm from the retromolar trigone, and 2 cm from the left corner of the mouth in relation to 34, 35, and 36. The surface of the lesion was irregular with indentation of teeth noted, and areas of clear fluid filled multiple small vesicles of varied sizes were seen 1.5 cm superiorly from the upper border of the lesion [Figure 2]. On palpation, the lesion was extending in submucosal plane and was not attached to the skin; soft in consistency, slightly tender, nonindurated, nonpulsatile and surrounding

mucosa appears normal. A left submandibular lymph node was palpable, measuring 1 cm × 1 cm, movable but nontender. A provisional diagnosis of traumatic hematoma was made with differential diagnosis of lymphangioma and hemangioma was given. After taking consent from the patient, surgical excision was planned. The patient underwent an uneventful excision under local anesthesia in proper aseptic conditions. No obvious feeder vessel was identified during surgery. The surgical site was closed primarily, and the excised specimen was sent for a histopathological examination. Gross specimen was soft in consistency and reddish in color and approximately of size 2 cm × 1.5 cm in dimension.

Hematoxyline and eosin stain section reveals para keratinized stratified squamous epithelium and underlying moderately collagenous connective tissue stroma. The connective tissue showed numerous dilated lymphatic's containing lymph. Many channels are also found with blood cells. Muscles fibers are seen deeper in the connective tissue stroma. Histopathological features suggestive of hemangiolympangioma was made [Figure 3]. The patient is on continuous follow-up and no recurrence was noted till date.

## DISCUSSION

Vascular anomalies are divided into two primary biological categories: vasoproliferative or vascular neoplasms and vascular malformation by International Society for the Study of Vascular Anomalies.<sup>[1]</sup> The key difference between the two types is whether there is increased or decrease endothelial cell turnover, based on histopathology.<sup>[4,10]</sup> Vasoproliferative neoplasms have increased endothelial cell turnover, while vascular malformations do not have



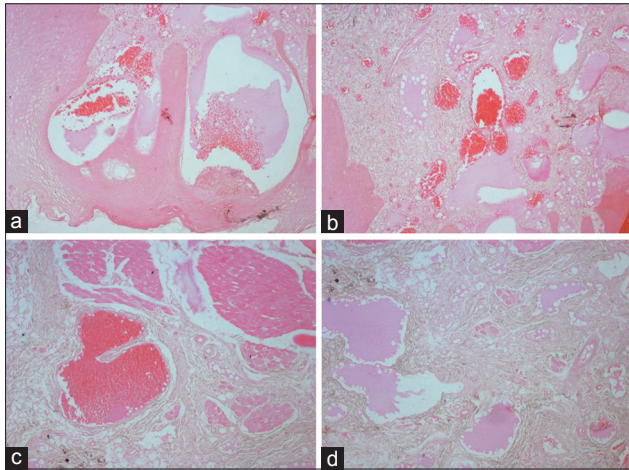
**Figure 1:** Clinical photograph of patient showing asymmetry of left cheek



**Figure 2:** Preoperative Intraoral picture of left buccal mucosa showing swelling and pinhead-sized, clear vesicles along with reddish exophytic masses

**Table 1: Case reports on hemangiolymphangioma published by different authors**

Author	Site	Sex	Age	Diagnosis	Year of study
Kim <sup>[7]</sup>	Left body of mandible	Male	62	Hemangiolymphangioma	2003
Jian <sup>[9]</sup>	Tongue	-	-	Lymphangiohemangioma	2005
Sobhana <i>et al.</i> <sup>[6]</sup>	Buccal mucosa	Male	18	Hemangiolymphangioma	2012
Hunchaisri <sup>[8]</sup>	Floor of mouth	Female	41	Hemangiolymphangioma	2013
Yarmand <i>et al.</i> <sup>[1]</sup>	Buccal mucosa	Male	23	Lymphangiohemangioma	2016
Selvamani M <i>et al.</i> (present study)	Buccal mucosa	Female	21	Hemangiolymphangioma	2017



**Figure 3:** (a and b) Photomicrograph (H & E, stain  $\times 10$ ) reveals stratified squamous epithelium with juxta epithelial stroma showing numerous vessels spaces which are filled with either blood cells or lymphatic fluids. (c) Photomicrograph (H & E, stain  $\times 10$ ) showing dilated vessels filled with RBC's along with few cross sections of muscle bundles. (d) Photomicrograph (H & E, stain  $\times 10$ ) showing numerous vessels spaces which are filled with lymphatic fluids

increased endothelial cell turnover.<sup>[3]</sup> Instead, vascular malformations are structural abnormalities of the veins, arteries, capillaries and lymphatic vessels.<sup>[3,11]</sup>

Most of the vascular anomalies are comprised Hemangiomas. Usually, seen in relation to the head and neck and are the most common tumors of infancy. Hemangiomas and vascular malformations may appear to be very similar but their course and treatment are different. Hemangiomas appear in the first few weeks of life, presents a rapid growth (proliferative) phase which usually lasts 6–10 months, followed by gradual involution, i.e., regress spontaneously over time.<sup>[12]</sup> Structurally, blood vessels are incomplete in hemangiomas, and they are surrounded by hyperplastic cells.<sup>[13]</sup> Vascular malformations are anomalies of blood vessels which present from birth even though they might not be apparent and persist throughout life.<sup>[12]</sup>

Vascular malformations do not show active proliferation, but they consist of ecstatic vessels of either one of the vein, capillaries, arteries, lymphatic vessels or combinations.<sup>[13]</sup> Lymphangiomas are an abnormality of the lymphatic system, which developed from sequestration of the primitive lymphatic cells. Vessels are capable of accumulating fluids.

On the other hand, vessels do not anastomose with bigger lymphatic vessels; therefore, the lymphatic blockage may lead to the cystic appearance of lymphangioma.<sup>[14]</sup>

Very few cases have been reported in regard to lymphangiohemangioma or hemangiolymphangioma [Table 1]. Yarmand *et al.* presented a case of 23-year-old male with a chief complaint of swelling of the buccal mucosa with clinical presentation was pebbly, vesicle-like and the so-called “frog-egg” or “tapioca-pudding” appearance which is similar to our case. Secondary hemorrhage into the lymphatic spaces may cause some of these “vesicles” to become purple.<sup>[1]</sup>

According to the clinical features, age and the location of the occurrence, pyogenic granuloma could be the differential diagnosis. Pyogenic granuloma can present with clinical appearances, ranging from a sessile lesion to an elevated mass. It can appear deep red to reddish-purple in color and present with wide clinical appearances, ranging from a sessile lesion to an elevated mass. In contrast, hemangioma has more plum, histiocytoid, endothelial cell proliferation without an acute inflammatory cell infiltrate.<sup>[15]</sup>

In the present case, histopathological features did not show inflammatory cell infiltration in the stroma, and the combination of both lymphatic vessels and capillary vessels filled with red blood cells concluded the diagnosis of hemangiolymphangioma.

## CONCLUSION

Hemangiolymphangioma is rare in the oral cavity, and only a few cases reported till date. All suspected cases should be appropriately evaluated to rule out bony, intracranial and systemic involvements. However, surgical excision is the mainstay of the treatment. The patient should be followed long term to rule out any recurrence.

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## Conflicts of interest

There are no conflicts of interest.

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